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Long-Term Outcomes of SDR in Spastic Cerebral Palsy Patients

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Aim. Long-term outcomes of selective dorsal rhizotomy (SDR) are not described in detail in the literature. The aim of this study was to systematize and evaluate long-term outcomes of SDR in cerebral palsy (CP) patients from various functional groups. **Material and methods.** Forty seven patients with spastic CPs were operated on. All patients underwent SDR of the L1—S1 roots under EMG control. In all cases, osteoplastic laminoplasty was used as an approach. Surgical treatment outcomes were evaluated using the Ashworth and GMFM 88 scales. The data were subjected to statistical analysis. The follow-up period ranged from 12 months to 7 years. **Results.** A significant reduction in spasticity from 4.34±0.53 points before surgery to 1.61±0.45 points after surgery (p<0.001) was observed in most cases. The changes in locomotor functions were maximal in the 3rd GMFM class, increasing from 48±4% points before operation to 52±6% points 12 months after operation (p<0.042). The changes in locomotor functions amounted to 2% in the 4th GMFM class and 1% in the 5th GMFM class. The best functional outcomes were obtained in children under 10 years of age. A positive correlation was found only between the percentage of cut roots and a decrease in spasticity (r=0.85). No correlation between the amount of cut roots and the changes in locomotor functions was observed. No spinal cord deformities were observed in the follow-up period. **Conclusion.** The degree of spasticity reduction due to SDR is directly dependent on the amount of cut roots. The functional result of SDR is affected not only by a decrease in spasticity but also by the functional status and age of the patient at the time of surgery. In all cases, osteoplastic laminoplasty should be used as an approach to prevent spinal cord deformities.

Keywords: *spasticity, cerebral palsy, selective dorsal rhizotomy.*

Selective dorsal rhizotomy (SDR) is a neurosurgical procedure performed mainly at the lumbar level for reducing abnormal muscle tone in patients with lower extremity spastic paraparesis. The primary beneficiaries of SDR are children with spastic cerebral palsy (CP) [1, 2]. Spasticity is a stretching velocity-dependent increase in tonic stretch reflexes, resulting from the loss of inhibition of motor neurons by the upper central nervous system structures [3]. Selective dorsal rhizotomy reduces excessive activity by partial cutting of some dorsal spinal roots comprising the myotatic fibers — the pathways terminating the stretch reflex and mediating spasticity [4]. A meta-analysis of short-term outcomes of rhizotomy in three randomized controlled trials was performed in 2002. SDR was shown to lead to a reduction in spasticity, increase in the range of movement, and improvement in motor functions in the short term period (6 months to 1 year) [5—7].

The literature does not describe in detail the long-term outcomes of rhizotomy, being mostly focused on complications, such as spinal deformities and pain. The study of long-term outcomes of rhizotomy since 2011 has led to the following conclusions:

1) the functional result can not be reliably evaluated because changes in the locomotor status are different in various groups (GMFM scale); therefore, the changes in functions should be evaluated separately for each group;

2) the functional outcome should be assessed separately in different age groups;

3) long-term complications of rhizotomy, such as spinal deformities, may not be associated with surgery because they are typical of CP patients in general [8].

A study of SDR outcomes, which was carried out at the Burdenko Neurosurgical Institute in 1996, confirmed a high efficacy of the surgery for reducing abnormal muscle tone in the long-term period [1]. However, the study included a small sample of patients and did not assess the patients using gross motor scales. Complications, such as progressive spinal deformities, were observed in the long-term period, which was probably associated with the use of laminectomy as an approach.

The aim of this study was to systematize and evaluate the long-term outcomes of selective dorsal rhizotomy in CP patients from different functional groups according to the GMFM scale.

Material and Methods

A total of 47 children with CP in the form of lower extremity spastic paraparesis and tetraparesis were studied (Table): 32 (68%) boys and 15 (32%) girls; the mean age was 7.4±6.1 years.

All patients before surgery underwent repeated courses of conservative and rehabilitation treatment with a low and unstable clinical effect. Before surgery, all the patients underwent botulinum toxin therapy for lower extremity muscle spasticity (2—6 injections). A clinical effect was absent in 16 patients; 31 patients had a partial clinical effect regressing during subsequent injections. In 5 cases, patients had already undergone orthopedic interventions for fixed contractures: achilloplasty and hamstring tendon lengthening.

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The patients were selected for surgical treatment based on a comprehensive examination with involvement of a neurologist, physiotherapist, orthopedist, and neurosurgeon. The examination protocol included evaluation of muscle tone using the Ashworth's scale, evaluation of the range of active and passive movements, video recording of gait, and evaluation of the locomotor status based on the Gross Motor Function Measure (GMFM-88) scale. The Ashworth's scale grades spasticity from 1 to 5, where 1 is normal muscle tone, and 5 is the maximum muscle tone when passive movement is almost impossible. The GMFM scale consists of 88 standard motor function tests. Each test is graded on a four-point scale, depending on a test performance level: 0 — cannot initiate task, 1 — initiates task, 2 — partially completes task, 3 — completes task. The total sum of scores is represented as a percentage. Patients with 20% or less are classified as class 5 (most severe form of cerebral palsy), 20 to 40% as class 4, 40 to 60% as class 3, 60 to 80% as class 2, and 80 to 100% as class 1.

Before surgery, all patients underwent brain MRI to evaluate hydrocephalus and exclude concomitant intracranial pathology as well as MRI of the lumbar spinal cord to identify the medullary cone level. A control examination was carried out 3, 6, and 12 months after surgery and thereafter each year.

Statistical processing of the data was performed using the SPSS Statictics 6.0 software. A correlation analysis using the Pearson correlation coefficient (r) was performed to evaluate the relationship between the amount of cut fascicles and the changes in spasticity and locomotor functions. Given the small sample size and absence of normal distribution in the Kolmogorov-Smirnov test, the Wilcoxon nonparametric test (T) was used to analyze the significant differences between preoperative and postoperative values of muscle tone and locomotor functions in different groups of patients.

The study included patients treated in the period from 2007 to 2014. Patients operated on later were not studied because short-term follow-up interfered with an adequate assessment of the changes in locomotor functions over time. The inclusion criteria were as follows:

1) high muscle tone (3 or more points on the Ashworth scale);

2) conservative therapy failure;

3) botulinum toxin therapy failure.

The contraindications for rhizotomy included:

1) secondary muscular dystonia;

2) hyperkinetic syndrome;

3) somatic contraindications.

Mental retardation was not considered to be a contraindication for rhizotomy.

All patients were operated on under general anesthesia. Before being transported to the operation room, patients were subjected to intramuscular premedication with ketamine and benzodiazepines according to anesthesiologist orders. At admission to the operation room, anesthesia induction, tracheal intubation, and mechanical ventilation were performed. All children underwent placement of a Foley urinary bladder catheter. Anesthesia was maintained with an infusion of propofol and fentanyl under monitoring the depth of anesthesia (BIS monitor). Since the operation was performed under EMG control, it was important to maintain the adequate depth of anesthesia in order to obtain true neurophysiological responses. The use of muscle relaxants was limited to intubation only. Electrodes for intraoperative myography were inserted into the adductor muscles, hamstring muscles, quadriceps and gastrocnemius muscles, and anal sphincter **(Fig. 1)**.

Osteoplastic laminotomy at the T12—L1 level was performed with the medullary cone being in the normal position. In 2 patients with a fixed spinal cord, laminotomy was performed at the L3—L4 level. After opening the dura mater, the medullary cone was identified. The L1 dorsal root was identified in the radicular foramen and was isolated to its branching from the dorsolateral sulcus of the spinal cord. Further, moving along the dorsolateral sulcus downward, the L2—S1 dorsal roots were successively isolated. Each root was divided into several root fascicles. The root fascicles were subjected to intraoperative electrostimulation using a mono- or bipolar stimulator (frequency of 5 Hz, pulse duration of 0.1—0.2 ms, amplitude of 0.5—2 mA). In this case, motor responses in the legs were evaluated. Responses occurring in different muscle groups or on the contralateral side were considered as abnormal. The fascicles producing these responses were coagulated and cut **(Fig. 2)**.

If no or low-amplitude motor responses were detected during stimulation of fascicles, the fascicles were left intact. In this case, if S1 root stimulation led to responses in the sphincter, the fascicles were left intact to prevent pelvic organ dysfunction. The S2 root was left intact in all cases. A total of 50 to 75% of the root amount was cut. When placing a bone block back in its original position, T12—L1 vertebral arches were cut; the spinous processes were cut to 2/3 of their length; and then, a vertebral arch was broken off to eliminate diastasis between the vertebral arches and the cut bone block.

After surgery, a urinary catheter was left for one day for prophylaxis of possible urinary retention. The patients were administered tramadol intramuscularly 3 times a day for 4 days to prevent pain in the postoperative period. Verticalization of patients was started on the 45th day. The patients were recommended wearing a lumbar corset when sitting and lying for 3 months. After this period, control spondylography was performed. In the absence of progressive deformities, the corset was removed. Physiotherapy was started after discharging patients from the hospital. The degree of residual flexion contractures was assessed three 3 months after surgery, and a decision on orthopedic surgical treatment was made.

Clinical cases

Patient	Age, years	GMFM before, %	GMFM after, %	Muscle tone before	Muscle tone after	Follow-up, years
1. *K.*	5	11	11	5.0	3.0	7
2. *Z.*	3	24	28	5.0	2.0	6
3. *G.*	6	25	28	4.0	1.0	6
4. *B.*	5	26	26	4.0	1.5	6
5. *R.*	3	10	10	4.5	1.5	6
6. *L.*	19	44	46	4.0	1.5	6
7. *D.*	14	30	35	4.0	2.0	6
8. *D.*	33	15	15	5.0	2.0	6
9. *R.*	4	21	25	4.5	1.5	6
10. *S.*	7	34	38	4.5	1.5	6
11. *B.*	4	15	18	4.0	1.0	5
12. *V.*	15	34	34	3.5	1.5	5
13. *B.*	5	10	10	5.0	1.5	5
14. *S.*	5	10	21	3.5	1.0	5
15. *A.*	6	35	35	5.0	2.0	5
16. *Yu.*	7	22	22	3.5	3.5	5
17. *P.*	7	30	30	5.0	2.0	5
18. *F.*	7	55	61	5.0	2.0	4
19. *I.*	4	38	40	4.0	1.5	4
20. *B.*	5	25	25	4.0	2.0	4
21. *F.*	30	35	37	5.0	1.5	4
22. *F.*	4	19	20	4.5	1.0	4
23. *P.*	7	27	32	4.0	1.5	4
24. *P.*	7	14	16	5.0	2.5	4
25. *Ts.*	3	23	23	5.0	1.5	4
26. *E.*	7	1	1	5.0	1.5	3
27. *K.*	5	47	47	4.0	1.5	3
28. *M.*	4	46	52	4.5	1.5	3
29. *K.*	10	46	50	4.0	1.5	3
30. *S.*	4	48	48	4.5	2.0	3
31. *G.*	6	30	30	4.0	1.5	3
32. *V.*	9	28	33	3.0	1.0	2
33. *R.*	4	30	37	4.0	1.5	2
34. *V.*	3	27	30	4.0	2.0	2
35. *K.*	3	37	42	4.0	1.0	2
36. *B.*	5	30	30	5.0	2.0	1
37. *M.*	5	10	10	5.0	2.0	1
38. *B.*	9	53	62	4.0	1.5	1
39. *G.*	3	12	12	5.0	1.5	1
40. *S.*	11	15	15	4.0	1.5	1
41. *Sh.*	8	35	40	4.0	1.5	1
42. *G.*	5	25	25	4.0	1.0	1
43. *S.*	7	16	16	4.5	3.0	1
44. *E.*	10	30	35	5.0	1.5	1
45. *T.*	4	25	25	4.0	2.0	1
46. *A.*	5	25	25	4.0	1.0	1
47. *M.*	6	26	26	4.0	1.5	1

Results

In all cases, a reduction in abnormal muscle tone in the adductor muscles, gastrocnemius muscles, and hamstring muscles from 4.34±0.53 preoperatively to 1.61±0.45 postoperatively ($p<0.001$), on average, was observed in the early postoperative period. An increase in the range of passive movements in the hip, knee, and ankle joints was observed in association with regression of myogenic contractures. However, no changes in locomotor functions were observed in the early postoperative period. In 4 cases, delayed urination for



Fig. 1. **Positioning of electrodes for intraoperative myography.**

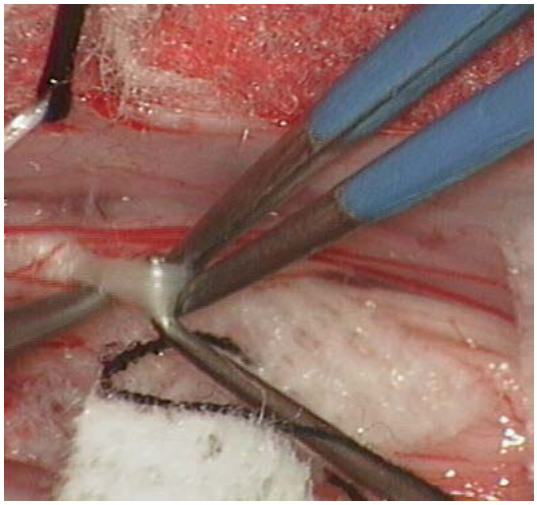


Fig. 2. **Coagulation of fascicles.**

2—3 days occurred in the early postoperative period. After this period, the disturbances resolved spontaneously.

An examination of patients after 3 months revealed an increased range of passive movements in all cases. In 21 (44%) cases, an increased range of active movements (flexion and extension in the knee and hip joints, increased dorsiflexion of the foot) was also observed. Contractures in the ankle and knee joints remained in 15 patients, which required additional orthopedic surgery on the lower extremities.

The changes in locomotor functions were observed 6 months after surgery. In the long-term follow-up (2 or more years after surgery), the total change in locomotor function was 3%: from 27±12% before surgery to 30±13% after surgery (p<0.009). The greatest change was observed in class 3 patients (GMFM scale) where it was 4%: from 48±4% before surgery to 52±6% after surgery (p<0.042). The changes in class 4 patients were less significant and amounted to 2%: from 29±5% before surgery to 31±6% after surgery (p<0.001). In class 5 patients, improvements were minimal — only 1%: from 12±4% before surgery to 13±5% after surgery (p<0.068) **(Fig. 3)**.

According to the Ashworth's scale, the changes in muscle tone in these subgroups were similar: 2.64 in class 3 (from 4.28±0.39 to 1.64±0.24); 2.84 in class 4 (from 4.61±0.5 to 1.77±0.69); 2.59 in class 5 (from 4.22±0.54 to 1.63±0.51) **(Fig. 4)**.

The best functional results were obtained in children under 10 years of age. The overall change in the locomotor status varied from 29±14% before surgery to 32±17% in the long-term period (p<0.057). The locomotor function status in three patients from this group was upgraded by 1 point (GMFM scale): 1 patient upgraded from class 5 to class 4; 2 patients were upgraded from class 3 to class 2. No significant changes in the locomotor status were observed in patients older than 14 years of age.

A analysis of correlation between the amount of cut root fascicles and the changes in muscle tone and locomotor functions revealed a direct relationship only between the percentage of cut roots and spasticity regression (r=0.85). No direct relationship between the amount of cut root fascicles and changes in the locomotor status was found.

Four cases of spastic syndrome recurrence were observed in the long-term period. In two cases, spasticity relapsed only in the gastrocnemius and adductor muscles and did not reached the preoperative level. The state of another two patients returned to the preoperative level. A persistent clinical effect was observed in the other cases.

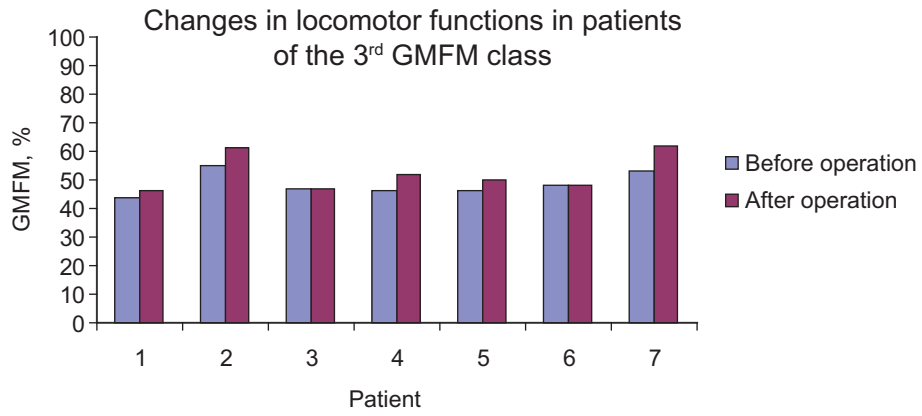
In one case, wound CSF leakage developed on the 3rd postoperative day. In this connection, a lumbar drain was used for 5 days, which resulted in elimination of the leakage; the wound healed by primary intention. There was no progression of spinal deformity in any case.

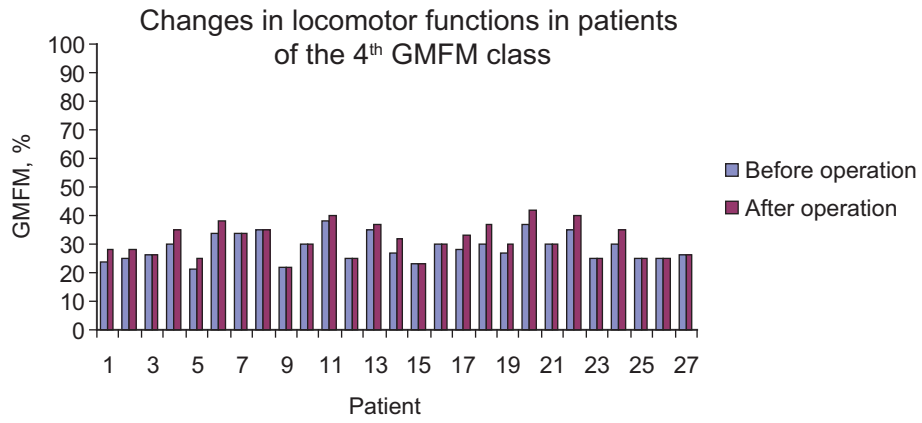
Discussion

Our findings demonstrate that selective dorsal rhizotomy is an effective technique for reducing severe spastic syndrome in CP patients. Spasticity resolves immediately after surgery, and the effect is persistent in most cases for many years [9, 10].

The study showed that the greatest changes in the locomotor status are typical of class 3 patients (GMFM scale). Similar results were reported in papers of other authors [11, 12].

The GMFM scale predicts the efficacy of planned surgery and defines the limits of outcomes that can be achieved in each particular case. The goal of treatment of class 5 patients is to facilitate the care for the patients and prevent gross deformities of the musculoskeletal system. In this group of patients, no significant changes in the locomotor status can be achieved; the child's parents should be first informed of this problem. Class 4 patients may develop positive changes in locomotor functions





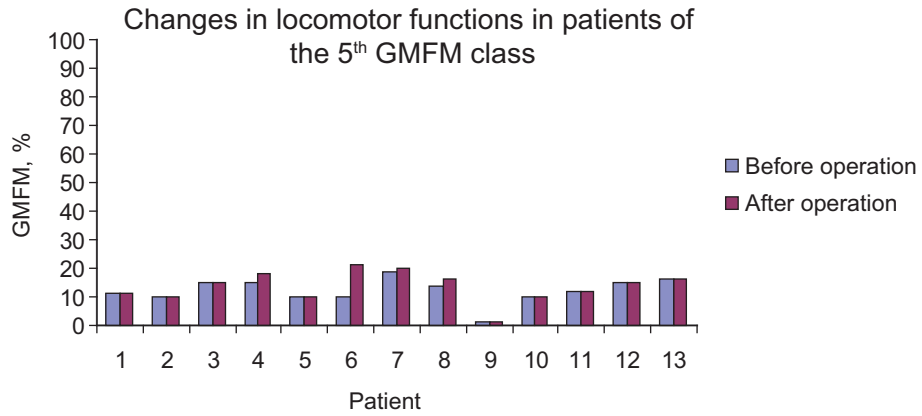


Fig. 3. **Changes in locomotor functions in patients of classes 3, 4, and 5 (GMFM scale).**

and learn new motor skills; unfortunately, the rehabilitation capabilities in this group of patients are limited. Class 3 (GMFM scale) has the most favorable prognosis. These patients will be capable of walking on their own [13, 14].

An important predictor of the surgery efficacy is the patient age. According to our findings, the greatest changes in the locomotor status were typical of patients under 10 years of age. The importance of early rhizotomy is also emphasized in a number of works by other authors. The preschool age, up to 7 years, is believed to be optimal [15—17]. Indeed, gross fixed contractures do not form at this age, and patients after correction of spastic syndrome will likely not require orthopedic interventions [18]. Furthermore, the movement pattern forms in this period, which makes rehabilitation treatment most effective. In patients older than 10 years of age, the changes in the locomotor status were less significant, while no changes occurred in patients older than 14 years of age. Therefore, rhizotomy in adolescent patients is palliative treatment

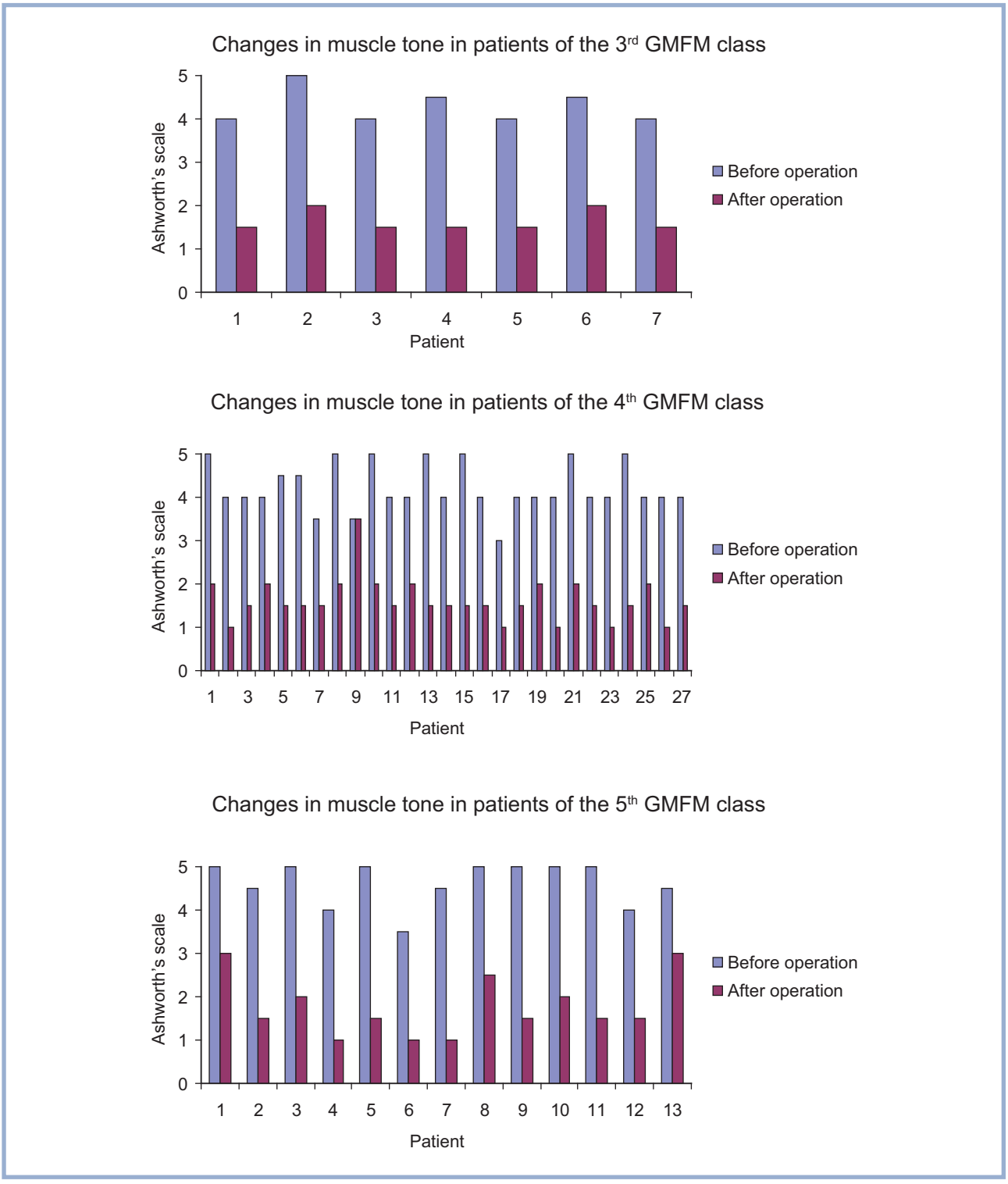


Fig. 4. **Changes in muscle tone in patients of classes 3, 4, and 5 on the GMFM scale.**

aimed just to facilitate care and reduce pain associated with spasticity [19].

Various studies report different data on the amount of sensory root resection, ranging from 50 to 75% [20, 21]. In this study, we analyzed a relationship between the amount of cut root fascicles and the changes in muscle tone and locomotor status. The amount of root resection was found to directly affect muscle tone. The greater the amount of cut root fascicles was, the larger the changes in spasticity were. This is quite logical because the greater the number of cut myotatic fibers is, the less the afferent activation of alpha motor neurons is. However, no direct relationship between the amount of cut roots and the changes in locomotor functions was found. This confirms

the fact that reduced spasticity is not the only requirement for improving motor skills. The disease severity, age of surgery, mental development level, rehabilitation treatment, and others factors also affect motor skills. However, an insufficient amount of cut fascicles leads to that spasticity may persist in some muscle groups and interfere with further rehabilitation. On the other hand, excessive resection of fascicles may result in muscle weakness, which will deteriorate the locomotor status of the patient. Therefore, it is important to determine the amount of dorsal root resection in each particular case. All the time, we relied on EMG control results: only those fascicles were cut that produced high amplitude responses in several muscle groups or bilateral motor responses. Fascicles producing low-amplitude responses or responses in one muscle group only were left intact. In the long-term period, we had 4 (8.5%) cases of spasticity recurrence. The mean amount of dorsal root resection in these patients was 60±3.56%, which was significantly lower than the mean amount of resection in the entire group — 69±5%. This suggests that a sufficient amount of fascicles should be cut for achieving a sustained clinical effect. The same conclusion is made in other studies on this topic. The amount of root resection in rhizotomy at the lumbar level should be at least 65%, otherwise spasticity is likely to relapse.

In contrast to a number of other studies [22—24], no further progression of spinal cord deformity was observed in patients from our series of operations. We believe this is associated with the used laminotomy technique. As an approach, we used osteoplastic laminotomy of the T12—L1 arches with mandatory preservation of the articular processes. In all cases, before placing the bone block back in its original position, the vertebral arches were cut to eliminate diastasis between the block and arches. Therefore, we fully preserved the structures of the posterior supporting complex of the spinal cord.

Conclusion

Selective dorsal rhizotomy is an effective technique for improving spasticity. The degree of a muscle tone reduction depends directly on the amount of cut fascicles. In addition to decreased muscle tone, the functional outcome of rhizotomy is also affected by the patient's baseline functional status and age of surgery. In any case, the operation should be performed under EMG control, and the amount of root resection should also be taken into account. Osteoplastic laminotomy should be used as an approach for preventing spinal cord deformities.

Authors declare no conflict of interest.

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Commentary

Various cerebral palsy (CP) forms are predominated by spastic palsies where lower extremity spastic paraparesis is the basic syndrome. So far, 20 to 42% of treatment outcomes in patients with normal intelligence remain unsatisfactory.

Management of abnormal muscle tone is the main problem in the treatment of CP patients. Functional neurosurgery techniques may be recommended for use at a certain stage if conservative treatments fail.

At present, selective dorsal rhizotomy at the level of dorsal lumbar and upper sacral roots, which was developed by W. Peacock and L. Arens in 1982, is most often used for treatment of patients with lower extremity spastic paraparesis. The operation involves separation of the dorsal roots into fascicular groups and cutting of those producing abnormal motor responses in the form of tonic muscle contraction during stimulation. At the same time, some issues associated with the use of this treatment technique still remain poorly understood. Most authors described not so much contraindications as conditions where the surgery effect was ambiguous and noted adverse effects of the surgery on locomotor functions of patients who used their spasticity for supporting or walking. Orthopedic complications associated with laminectomy have been insufficiently covered in the literature; there are only a few mentions of such a possibility.

Having studied long-term outcomes by means of the GMFM 88 comprehensive evaluation scale, the authors determined the surgery effect in terms of locomotor function preservation and spasticity reduction. The article describes in detail the surgical technique and methodology of neuromonitoring. The authors pay special attention to the problem of spinal deformities at the thoracolumbar junction as well as to the methods of laminoplasty and preservation of the intervertebral joints for the stability purpose. In conclusion, the authors summarize up the results of work and provide recommendations for the optimal age of the intervention, amount of fascicle resection, and expected time of surgery-based improvements.

The work used modern methods of statistical analysis of a significant amount of the international literature data. However, the authors did not mention a study by A.Yu. Stepanenko (1996), whose thesis topic is the same as the article title. That study carried out at the same institution 20 years ago contains recommendations for the optimal age of the operation (6—10 years), describes a relationship between the amount of cut fascicles (at least 65%) and the spasticity reduction degree, and covers the issues of spinal stability preservation in these interventions. The work of 1996 was based on technologies used in selective dorsal rhizotomy that were put into 3 inventions and innovations. In general, the efficacy of this technique achieves similar levels — 27—40%.

It should be noted that methods of functional surgery, being a part of comprehensive treatment of children with severe cerebral palsy, significantly improve the patient's quality of life and require consolidation of knowledge and methods of surgery in terms of experience accumulation and analysis of the results of foreign and domestic research.

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